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Full Length Research Paper

Paediatric mucoepidermoid carcinoma of palate: Literature review and report of a case

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Mucoepidermoid Carcinoma (MEC) is the most common malignant tumor in salivary glands. About 70% of these tumors are found in the parotid gland, 15 to 20% in the oral cavity, mostly in the palate, and 6 to 10% in the submandibular gland. MEC has female predilection and the mean age at onset is in the 5th decade of life. The treatment decision depends on the histopathology (Low-grade, Intermediate-grade, High-grade). A 12 year-old male with a right palatal tumor, having no association with vital teeth arrived at our service centre. The excisional biopsy reported was: MEC Intermediate-grade. Because few cases of MEC in Paediatric patients have been reported, a review of the literature, with range of age 0-17 years, was made, 24 patients with diagnosis of MEC in Palate, included our case, were found, the average age was 14 (29%), the predilection in genre was female with 19 cases (79%), and 5 cases in Male (21%). MEC has a female predilection and is uncommon in the first decade of life but should be considered even in paediatric patients. The prognosis depends on the clinical stage, grade of the tumor, location, and adequacy of treatment. The presence of nodal disease and distant metastases implies a poor prognosis.

Key words: Mucoepidermoid carcinoma, palate, childhood.

INTRODUCTION

Mucoepidermoid carcinoma (MEC) is the most common malignant salivary gland tumor, statical data indicate that it is the most common parotid malignancy (89%) and that is the most common malignant salivary gland tumor in children. About 70% of MEC are found in parotid gland, 15-20% in oral cavity, mostly in the palate, and 6 to 10% in submandibular gland. MEC has female predilection 3:1 at fifth decade of life (Marx and Stern, 2012). MEC was originally described by Volkmann in 1895 (Bhat et al., 2014), the oral cancer foundation refers that MEC was

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described by Masson and Berger in 1924 and in 1945 Stewart recognized MEC of salivary glands as a separate entity among salivary neoplasm (Ranganath et al, 2011).

Treatment decisions are based on histologic grading. The grading of MEC depends on the mix of cell types. Low grade is characterized by well-form cyst that contains mucin, lined by a mixture of mucos, intermediate and epidermoid cells. Intermediate grade contains smaller and fewer cyst and have more solid appearance, consist of intermediate cells, epidermoid cells, and some mucous cells.

High grade are solid and consist of intermediate and epidermoid cells, which show considerable atypia and mitotic activity is present (Marx and Stern, 2012). A review of literature about MEC of Palate revealed 24 documented cases in paediatric patients, included a case reported by our service of Oral and Maxillofacial Surgery at School of Dentistry of Universidad Autónoma de Nuevo León, México.

Case presentation

A Twelve-years-old Male arrived to our service; a complete anamnesis was made, the patient does not have antecedents or family cancer history, the only relevant antecedent was previous dental injury in anterior- inferior teeth 6 years before. He refers began with a mass in Right Palate five months previous (May, 2015), which was detected by his dentist, biopsy by puncture was practice that time without specific diagnosis.

At clinical examination was made, with oval face, esqueletal class III, abscense of inferior anterior theet, tongue and the rest of tissue had normal characteristics, a mass in the right hard palate, 2 cm in size was observed, with blue/red color, diffuse borders, fluctuant, and sesil base, the cervical palpation does not reveal lymph nodes (Figure 1). A Cone Beam was taken, without any bone involvement. An Incisional Biopsy was made (October, 2015), because the characteristics of clinical presentation, it was made superficial and the histopathology report us normal tissue. It was decided to excisional biopsy with a scalpel No. 15, removing the entire lesion 0.5 mm around, hemostasis was made with electrocautery, and an obturator for the defect of tissue was used.

The histopathologic analysis, made with hematoxilyne-and-eosin by the Pathology Department, reported presence of quistic sppaces covered by epidermoid cells and mucus secreting cells. Filling these spaces are nests and diffuse layers of these same cells between a fibrous connective tissue, which correspond to MEC of intermediate grade (Figure 2). The patient actually, 9 months after, has no evidence of lesion; a new Cone beam was taken, with no evidence of bone involvement. An oncological review was conducted by the specialist with no evidence of tumor.

RESULTS

The literature review include search in Pub Med, JOMS and IJOMS, the range of age was 0 to 17 years, 24 patients with diagnosis of MEC in Palate, including our case, were found. The youngest age was 5 years; 1 case (4%), the oldest 17 years old; 3 cases (13%), the average of age was 14 (29%). The predilection of genre was female with 19 cases (79%), and 5 cases in Male (21%) (Table 1). The most common location was hard palate; 17 cases (71%), followed by junction of hard and soft palate; 4 cases (17%), and 3 cases of soft palate (12%). The time of evolution was not specified in almost all the cases, but 1 year (9%) and 1 month (9%) was the most common. The size was not specified in almost all the cases 13 (54%) and 3 patients reported 2 cm (13%) (Table 1). One case reported bone involvement and another reported erosion of bone. Concerning the Histologic grade: low grade was the most prevalent with 22 cases (92%) of the total, 1 intermediate (4%) and 1 was not specified (4%). The common treatment was excision. One case had recurrence on 5 years follow up (Table 1).

DISCUSSION

The firm attachment of the mucosa of hard palate to the underlying periosteum, the anatomic proximity of the mucosa to the bone, and the abundance of minor salivary glands make the hard palate a unique anatomic region. Within this context, neoplastic lesions of the hard palate may show different characteristics and different histopathologic compositions compared with other anatomic regions. (Aydil et al., 2014). MEC is actually an uncommon tumor in minor glands in child. About 1% - 5% of all salivary glands tumors develop in children and adolescents, and the MEC are the most common malignancy. (Ritwik et al., 2012). In 1987 only 17 cases of MEC of the palate in children had been published. (Gustafsson et al., 1987). Although this salivary gland tumor represents a rare oral lesión in children, it should be considered when a lesion has a similar appearance as a mucocele but is found at a site other than the lower labial mucosa. (Flaitz, 2000). The mucocele is a common, reactive lesion of salivary glands that is seen frequently in children. This lesion presents as a dome shaped, translucent blue swelling that is fluid-filled. (Flaitz, 2000). Few series of cases describing salivary gland tumors in the pediatric population have been published. This Case represent an additional case of a Intermediate-grade MEC of the palate of a 12 years old child. Here, the average of age was 14 (29%), the predilection of genre was female with 19 cases (79%),

Figure 1. Initial clinical presentation

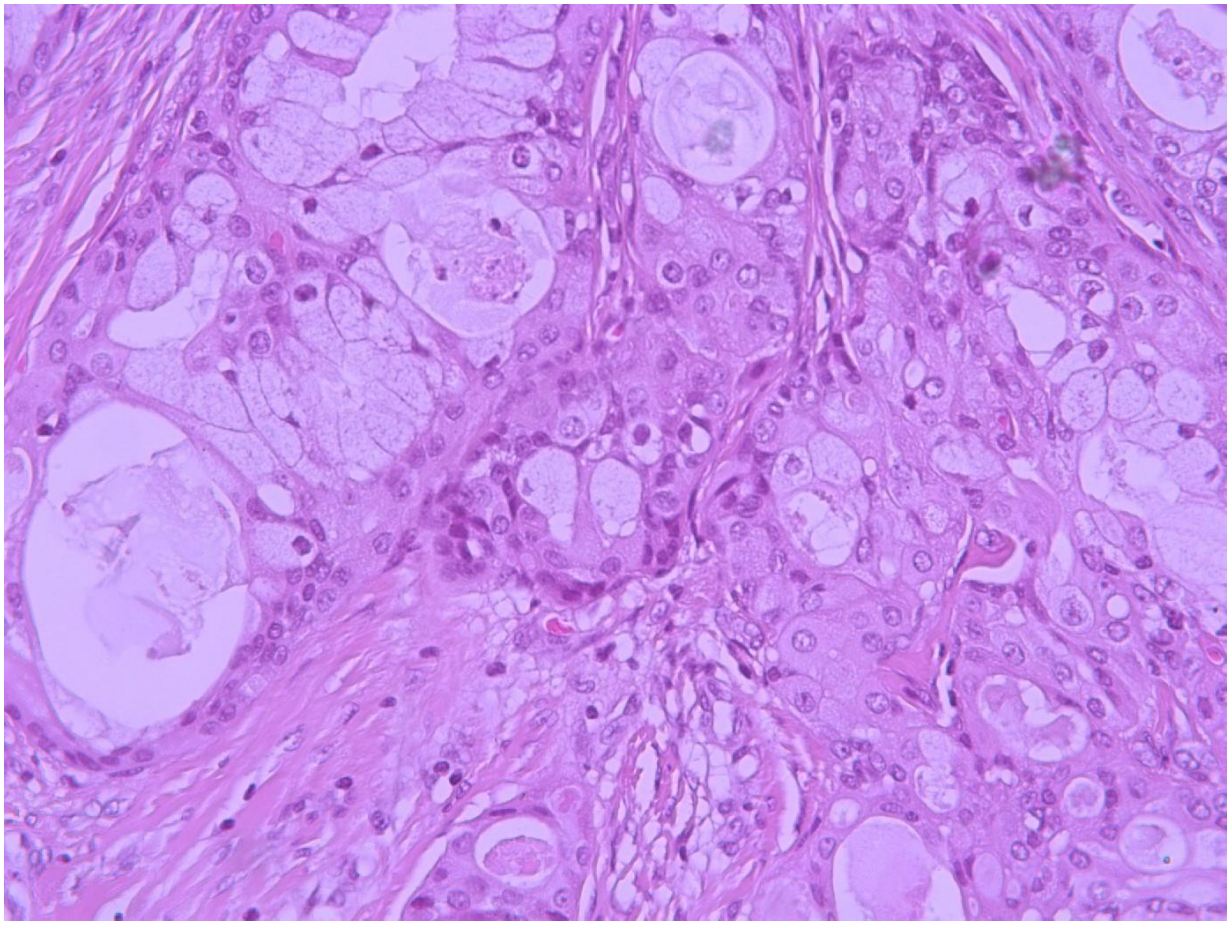

Figure 2. Photomicrograph of hematoxylin-and-eosin section of MEC. 40X Magnification.

Table 1. Paedriatric MEC reported at palate.

S/N	References	Case reported at the reference	Age	Genere	Site	Duration	Size	Bone involvement	Histological grade	Treatment	Recurrence	Follow-Up
1	Budnick (1982)	1	12	Female	Hard Palate	NS	2	NS	Low	Excision down to bone	No	3 years
2	Gustafsson et al (1987)	1	13	Female	Hard Palate	2 Months	1.5	NS	Low	Resección	No	1.5 years
3	Fonseca et al. (1991)	2	14	Male	Soft Palate	NS	NS	NO	Low	Two surgery	Yes/5 years	NS
4	Fonseca et al. (1991)	2	14	Female	Soft Palate	NS	NS	NO	Low	surgey	no	6 Months
5	April et al. (1997)	1	10	Female	Junction Hard/soft Palate	1 month	0.5	NO	Intermediate	NS	NS	NS
6	Aguilar et al. (1998)	1	13	Female	Junction Hard/soft Palate	7 months	3	NO	NS	Excision	NS	NS
7	Flaitz (2000)	1	8	Male	Hard Palate	9 monts	2	NO	Low	Wide local excision down to periosteum	NS	NS
8	Caccamese and Ord (2002)	23	17	Male	Junction Hard/soft Palate	NS	1.5	NO	Low	Local Resection	No	94 months
9	Caccamese and Ord (2002)	23	14	Female	Hard Palate	1 month	1	NS	Low	Excision	No	62 months
10	Caccamese and Ord (2002)	23	16	Female	Palate	1 year	2.5	NO	Low	Local Resection	No	42 months
11	Morales et al (2007)	1	14	Female	Hard Palate	1 year	5	NO	Low	Transoral resection	No	4 years
12	Pérez et al. (2008)	9	6	Female	Hard Palate	2 monts	NS	NS	Low	Local Excision	NS	NS
13	Pérez et al. (2008)	9	12	Female	Hard Palate	1 month	NS	NS	Low	Local Excision	NS	NS
14	Pérez et al. (2008)	9	14	Female	Hard Palate	4 months	NS	NS	Low	Local Excision	NS	NS
15	Pérez et al. (2008)	9	16	Male	Hard Palate	2 months	NS	NS	Low	Local Excision	NS	NS
16	Pérez et al. (2008)	9	17	Female	Hard Palate	2 months	NS	NS	Low	Local Excision	NS	NS
17	Pérez et al. (2008)	9	17	Female	Soft Palate	36 months	NS	NS	Low	Local Excision	NS	NS
18	Ávila et al. (2011)	13	16	Female	Palate	NS	NS	NS	Low	NS	NS	NS
19	Ritwik et al. (2012)	15	15	Female	Hard Palate	3 weeks	NS	NO	Low	Excision	No	21 years
20	Ritwik et al. (2012)	15	15	Female	Hard Palate	NS	3	NS	Low	Exicision	No	19 years
21	Ritwik et al. (2012)	15	14	Female	Hard Palate	NS	NS	NS	Low	NS	No	9 years
22	Sengul et al.(2013)	15	12	Female	Hard Palate	2 years	8 mm	Yes	Low	Excision	NS	3 years
23	Baumgardt et al (2014)	1	5	Female	Hard Palate	1 week	2.5	No	Low	Resection long the adjacent bone	NS	NS
24	This Case	1	12	Male	Junction Hard/soft Palate	5 months	2	No	Low	Excision	No	9 months

*NS: Not Specified.

the histopathological grade of prevalence was low grade, with 22 cases (92%). Fluctuance and a light blue color are helpful diagnostic clinical clues. MEC must be considered in the differential diagnosis of a lump or mass in a salivary gland-bearing area, especially the palate (Ritwik et al., 2012). Mucoepidermoid carcinoma requires surgical excision. Low-grade lesions may be treated with wide local re-section.

High-grade neoplasms often require treatment of the cervical lymph nodes by a selective neck dissection, radiation, or both (Ritwik et al., 2012).

Conflict of Interests

The authors have not declared any conflict of interests.

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